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Case Report

A CASE REPORT OF SPONTANEOUS BILOMA - AN ENIGMATIC SURGICAL PROBLEM

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ABSTRACT
Occurrence of biloma is a rare surgical condition often caused by surgery, interventional procedures and abdominal trauma. Spontaneous biloma is even rarer. A 38 year old otherwise healthy male, without any recent past history of surgery, intervention or trauma presented with biloma and recurred twice after treatment by both open surgical and percutaneous drainage, along with appropriate endoscopic biliary intervention, presented a surgical challenge. The reported case describes the shortcomings of current management of spontaneously occurring biloma and emphasizes the need for longer duration of placement for the percutaneous drainage to avoid recurrence of this enigmatic surgical condition.

Keywords: Biloma, Management

INTRODUCTION
Biloma is defined as bile collection, either encapsulated or not, outside the biliary tree, with intra- or extrahepatic location, generally of iatrogenic nature or results from abdominal trauma (Akhtar *et al.,* 2007) & (Bas *et al.,* 2011). The word "biloma" was first utilized by Gould & Patel, in 1979 (Kannan *et al.,* 2009), but there are descriptions of such an entity since the century XIX (Kaushik *et al.,* 2004*)*.
Spontaneous rupture of the biliary tree is rarely observed, sometimes being associated with choledocolithiasis (Bas *et al.,* 2011) & (Lee *et al.,* 2007*)*. The detergent activity of bile acids provokes chronic inflammation that, in turn, causes adhesions, leading to a possible loculated appearance of the collection (Akhtar *et al.,* 2007). Clinically, abdominal pain, distention, peritonitis, jaundice and, in more severe cases, sepsis, may occur (Akhtar *et al.,* 2007) & (Lee *et al.,* 2007). The mean time between onset of symptoms and the diagnosis is one to two weeks (Akhtar *et al.,* 2007). Considering the rarity of such condition, the authors describe the present case and review this rare clinical entity.

CASES
A 39 year old previously healthy male admitted in a medical ward with a clinical diagnosis of ascites. He had low grade fever and mild abdominal pain for two weeks before admission. On clinical examination, he had mild fever of 99 degrees of F along with abdominal distension. There was clinically demonstrable fluid thrill but shifting dullness was absent. Features of peritonitis were absent. Laboratory test results included: Total Leucocyte Count 24200 with polymorphs 92% gamma-GT: 55 U/L, Alkaline phosphatase 164U/L direct bilirubin levels: 1.4 mg/dl, and lipase levels: 49 U/L. Ultrasound scan showed intra-abdominal fluid collection, and a diagnostic aspiration was done which yielded thick greenish aspirate. Microbiological study of the aspirate presented negative results regarding culture, gram and Z-N staining. Ultrasonography (US) demonstrated encysted collection at Right Sub diaphragmatic, Right hypochondrium & left lower abdomen with internal echoes & septations. Biliary tree was normal except thick walled Gallbladder (Figure1). Computed tomography (CT) confirmed presence of perihepatic collection with foci of air (Figure 2) but normal biliary anatomy. Hollow viscous perforation was ruled out by an erect Chest X-Ray. Subsequent to transfer under Surgery the patient was submitted to laparotomy by a midline incision, as his fever and abdominal tenderness increased despite antibiotic therapy, which confirmed the tomographic description of abdominal collection of infected bile in sub diaphragmatic and sub hepatic region, infra colic compartment including pelvis was free of collection as shown in the Computerized Tomography (CT) scan (Figure3). Per operative examination of bowel, gallbladder and bile duct were normal, however intra operative cholangiogram could not be done for

technical reason. After thorough saline lavage two wide bore tube drains were placed in right hypochondrium and pelvis, which were removed after 7 days when they stopped draining. Patient was discharged on 10th day.

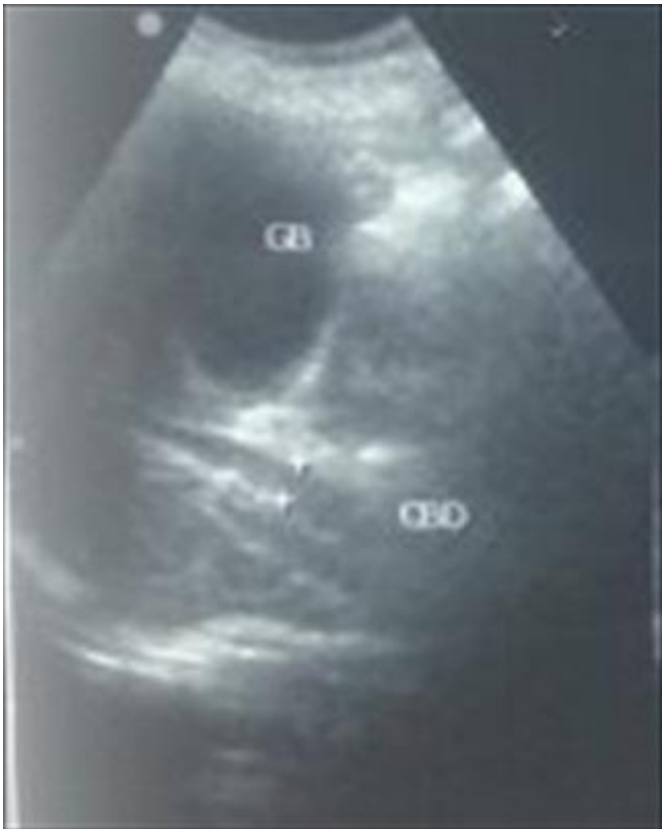

Figure 1: Ultrasound scan showing thick walled gallbladder with normal biliary anatomy

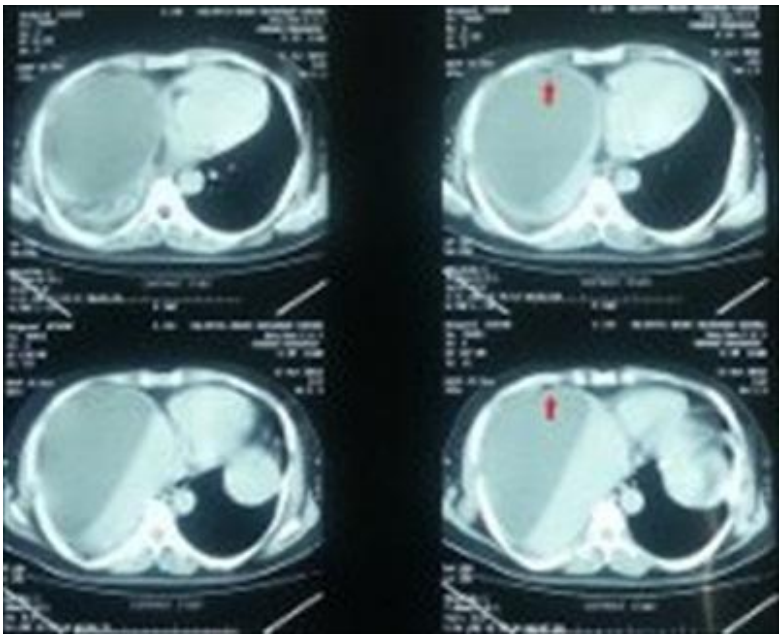

Figure 2: CT scan showing peri hepatic collection with air inside, marked with red arrow

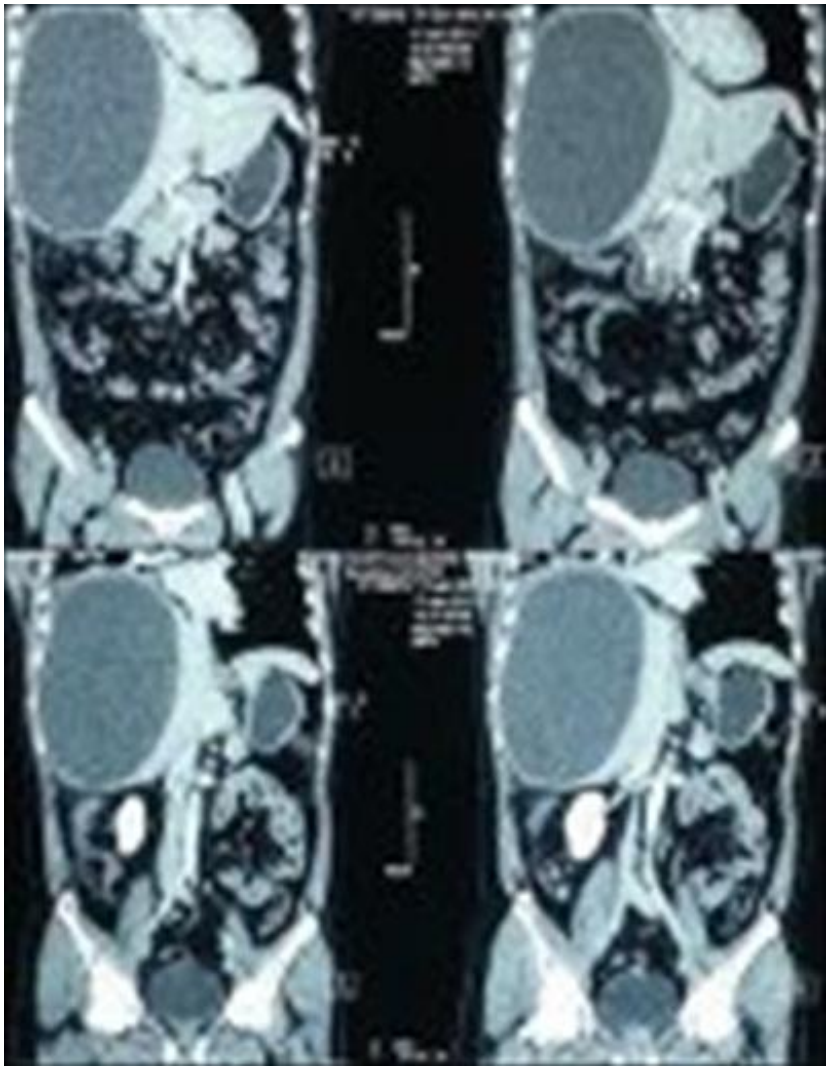

Figure 3: CT scan shows infra-colic compartment free of collection

But after about 2 months the patient again presented to us with recurrence of collection in Right Hypochondrium measuring 11.5* 16 cm. He had deranged Liver Function Test (LFT) with threefold increase of Alkaline phosphatase and dilated Common Bile Duct (CBD) on U/S scan. A CT Scan & CT guided pigtail catheter drainage of biloma was done followed by an Endoscopic Retrograde Cholangio Pancreatography (ERCP).

On ERCP a small bile duct stone was removed & ductal stenting was done. After 2 weeks catheter drain output was minimal and thus removed before discharging the patient with advice for cholecystectomy after 4 weeks.

To our utter surprise the patient returned to us after a month with a localized swelling in the left iliac fossa region, CT scan revealed collection both in the supracolic compartment (Figure 4) and infra-colic compartment (Figure 5).

He had no pain abdomen or any other constitutional symptoms. A repeat pig tail catheter was placed within the collection and kept for 12 weeks, when we again evaluated the patient with the percutaneous drain stopped draining in the last 10 weeks and sonography revealing no recurrence of collection, laparoscopic cholecystectomy was performed and the biliary stent was removed 2 weeks post operatively. Patient is in follow up for two years without any further recurrence of collection or any adverse symptoms.

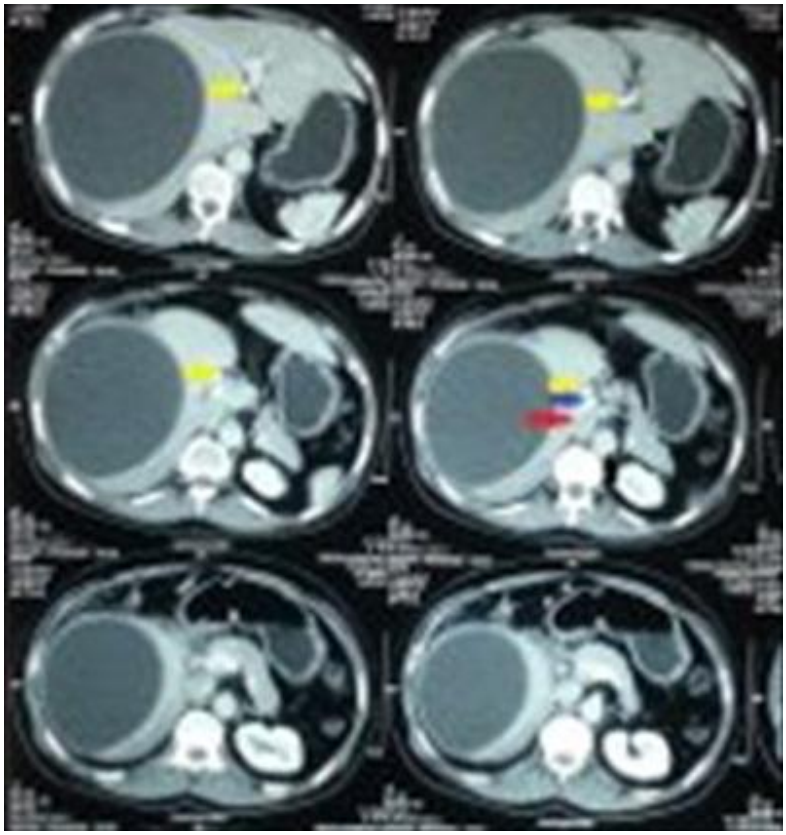

Figure 4: 2nd CT scan shows accumulation of bile in the supracolic compartment. Yellow arrow points to biliary plastic stent, blue arrow shows portal vein and red arrow the inferior vena cava

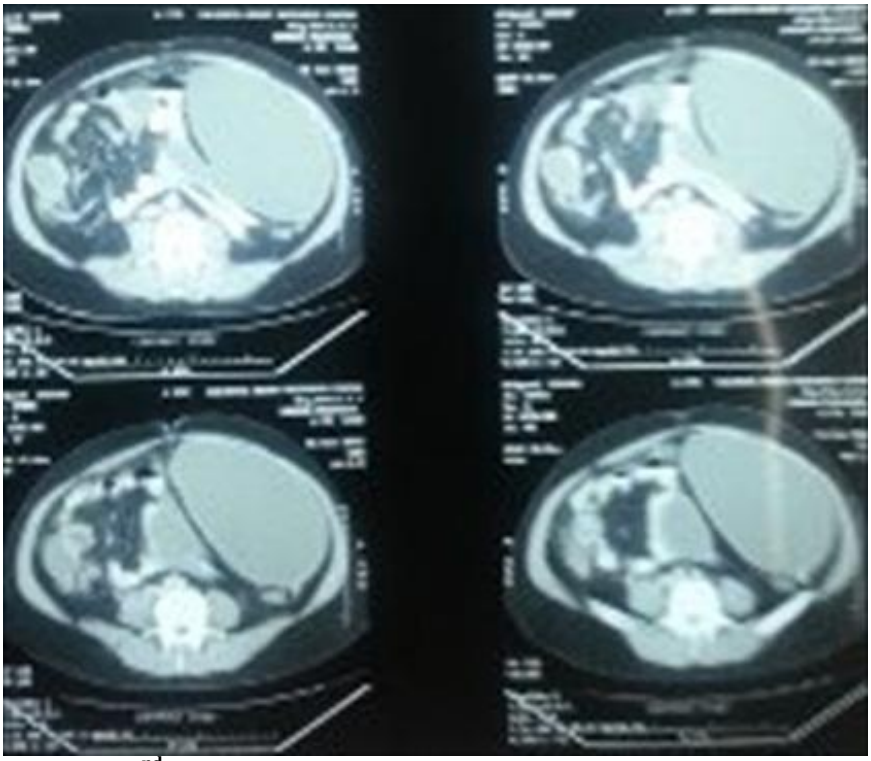

Figure 5: 2ndCT scan shows fluid in the infra-colic compartment also

DISCUSSION

Biloma formation is encountered mainly after surgical or interventional procedures and trauma involving the biliary system (Trivedi *et al.,* 2009) & (Yasar *et al.,* 2009). However, there are few reported cases of spontaneous biloma in the literature. The most frequent cause of spontaneous biloma is choledocholithiasis. Less commonly reported causes include biliary tree malignancy, acute cholecystitis, hepatic infarction and abscess, obstructive jaundice and tuberculosis. Although the pathophysiology of spontaneous biloma remains to be elucidated, one suggested contributing factor is increased intra ductal pressure due to obstruction on any part of the biliary tree. Bilomas are generally localized in the right upper quadrant of the abdomen, neighboring the right hepatic lobe. The clinical presentation of biloma varies greatly from nonspecific abdominal pain to biliary sepsis. Encapsulation of bile within the omentum and mesentery prevents generalized peritonitis in most cases. Abdominal US is the first modality to evaluate the nature of a biloma and the underlying pathology. However, an abdominal CT can define the disease, the cause and the relations with the adjacent structures more accurately. Differential diagnosis should include hematoma, seroma, liver abscess, cysts, pseudo cysts, and lymphocele. Percutaneous aspiration under radiologic guidance can also aid in diagnosis and treatment. Biochemical and microbiological analysis of the fluid helps differentiation from pyogenic abscesses or other causes. A Magnetic Resonance Imaging (MRI) may be of value to evaluate the etiology since it can be used safely for the pathologies of the biliary system. ERCP is also used for diagnostic and therapeutic purposes. Although some bilomas, especially those that are small in size and asymptomatic, can be followed up without intervention, most require interventional treatment. Percutaneous and endoscopic modalities provide adequate drainage and may be therapeutic in most cases. These treatments are preferable to surgery as the first step. ERCP is indicated, particularly in treatment failure, such as persistent bile leak despite percutaneous catheterization. Surgery always remains an option in emergency and persistent cases. In our patient, the biloma was located in the right upper quadrant and left lower quadrant with extension into pelvis & was detected with abdominal US & US guided aspiration. Because repeated US & CT Scan demonstrated normal biliary tree with normal LFT & patient developed sepsis the initial management was surgical drainage of infected biloma. Although the drain stopped draining after about 2 weeks & US showed no collection post-operative period, the patient again presented in outpatient after 2 months with recurrence in right upper abdomen and altered LFT. The biloma was drained percutaneously and an ERCP was done to rule out ductal pathology, to reduce ductal pressure & to find the possible site of leak if any. On ERCP a small CBD stone was removed & stent placed but it fails to identify the site of bile leak. Premature removal of the pigtail catheter resulted in recurrence of biloma necessitating repeat placement of pigtail catheter and kept in situ for 12 weeks which resulted in natural sealing of unidentified site of bile leak. Subsequently we performed Laparoscopic Cholecystectomy followed by biliary stent removal.

Conclusion

Spontaneous biloma is a rare occurrence which can deceive a clinician in diagnosis. A gamut of endoscopic and radiological investigations often fails to localize the site of bile leak. Definitive treatment of biliary pathology along with percutaneous drainage of biloma is the treatment, but the percutaneous drain should be kept in situ for a much longer time even when it stops draining any significant amount; as premature removal often results in recurrence. This leaves us with a dilemma –what shall be the treatment if the biloma recurs even after long term placement of the pigtail drain. Available literature does not answer the question clearly.

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